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A CASE OF RETROPERITONEAL BRONCHOGENIC CYST TREATED BY LAPAROSCOPIC SURGERY

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We describe herein a rare case of a retroperitoneal bronchogenic cyst successfully treated by laparoscopic surgery. A 39-year-old man with low-grade fever was referred to our hospital because of suspicion of an adrenal tumor. Abdominal computerized tomography (CT) and ultrasonography revealed a homogenous solid mass, 35×30 mm in diameter, in the left suprarenal region. Laboratory studies showed that the levels of adrenal hormones were normal except for the white blood cell count of 9,700/ μ L and C-reactive protein of 1.7 mg/dl. We diagnosed it as a non-functioning adrenocortical adenoma or an adrenal cyst. However, one year later he underwent laparoscopic surgery because the mass had gradually increased by 10 mm and the low-grade fever persisted. Pathological evaluation of the surgical specimens established the diagnosis of retroperitoneal bronchogenic cyst. The low-grade fever disappeared after the surgery.

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Key words : Bronchogenic cyst, Retroperitoneal space, Laparoscopic surgery

INTRODUCTION

Bronchogenic cysts in the retroperitoneal space are a rare developmental anomaly of the primitive foregut that usually develops in the posterior part of the mediastinum. To our knowledge, 42 cases of isolated retroperitoneal bronchogenic cysts have been described in the world, and the majority of bronchogenic cysts were removed by open surgery¹⁻¹⁰. Recently, laparoscopic and retroperitoneoscopic procedures have become widely used to treat such retroperitoneal diseases as functioning adrenocortical adenomas and relatively small adrenal pheochromocytomas. We report herein a man with a retroperitoneal bronchogenic cyst successfully treated using laparoscopic procedures.

CASE REPORT

A 39-year-old man was referred to our hospital in May 2003 for a left retroperitoneal mass suspiciously derived from the adrenal gland and a low-grade fever. The concentrations of catecholamines and their metabolites in both the plasma and urine, and the levels of plasma adrenocortical hormones, such as aldosterone and cortisol, were within the normal range while the inflammatory laboratory data showed a white blood cell count of 9,700/ μ L and C-reactive protein of 1.7 mg/dl. Abdominal computerized tomography (CT) and ultrasonography revealed a homogenous solid mass, 35 mm in diameter with a partial enhancement on the margin in the left suprarenal region. Iodine-131 MIBG and Adosterol scintigraphies did not demonstrate any finding of abnormal uptake. He was thus diagnosed as having a non-functioning adrenocortical adenoma or an infected adrenal cyst and kept on observation with a serial abdominal CT and the measurement of adrenal



Fig. 1. Abdominal CT revealed a 35 mm homogenous mass in the left suprarenal region (arrow).

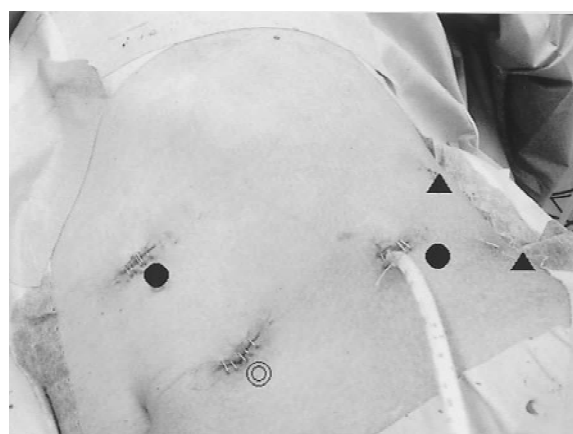


Fig. 2. The locations at which trocars were inserted for transperitoneal approaches. \odot : 12 mm trocar (for the endoscope); \bullet : 10 mm trocar; \blacktriangle : 5 mm trocar.

hormone activities every six months. However, the solid mass increased in size from 35 to 45 mm during the one-year follow-up (Fig. 1) and the low-grade fever persisted. In September 2004 he was admitted to our hospital and underwent transperitoneal laparoscopic surgery to remove the solid mass because of the possibility of the cause of slight fever and the suspicion of malignancy. At operation five laparoscopic trocars were inserted into the peritoneal space (Fig. 2). The anterior aspect of the left kidney was exposed after dissecting the peritoneum from the anterior lobe of Gerota's fascia. Then, the left renal vein and adrenal vein were dissected and the left adrenal gland was identified but the tumor was apparently isolated from the adrenal gland. The tumor was adherent to the peri-adrenal adipose tissue, and especially aorta and diaphragm in the retroperitoneal space. It was carefully dissected and removed en bloc. The operation time and blood loss were 144 minutes and 84ml, respectively. The tumor was 23 g in weight, and contained brown and slightly turbid liquid. The result of the culture inspection of the content liquid was negative. Pathological evaluation of the surgical specimen revealed a cyst lined with pseudostratified columnar epithelium and the cyst wall contained mucous glands corresponding to the mucosa of the bronchi and hyaline cartilage (Fig. 3). Thus, it was ultimately diagnosed as a bronchogenic cyst. The patient had an uneventful postoperative course and the fever was promptly alleviated.

DISCUSSION

A bronchogenic cyst is a congenital anomaly usually developing in the mediastinal space or lung. It stems from the pinching off of an irregular lung budding of the primitive foregut, with aberrant migration into the

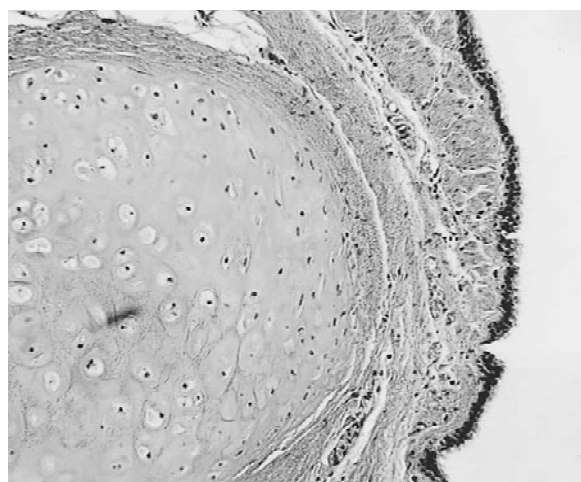


Fig. 3. Microscopically, the cyst was lined with pseudostratified columnar epithelium; the cyst wall contained mucous glands corresponding to mucosa of the bronchi and hyaline cartilage (hematoxylin and eosin stain; original magnification $\times 200$).

abdomen before fusion of the diaphragm during embryonal development.

To our knowledge there have been only 2 cases diagnosed as retroperitoneal bronchogenic cyst preoperatively^{2,5)}, and 94.7% of available retroperitoneal bronchogenic cysts (38 cases) were postoperatively identified by the pathological evaluation. The definitive pre-operative diagnosis of retroperitoneal bronchogenic cyst seems to be difficult because the majority of retroperitoneal bronchogenic cysts located peri-adrenal gland and no enhancement on CT scan like adrenal cysts. In addition, a bronchogenic cyst does not necessarily reveal typical cystic patterns when accompanied by inflammation or hemorrhage^{7,8)}. In the present case the CT scan did not reveal a typical cystic pattern because of the partial enhancement of the margin and the turbid liquid in the cyst suggesting inflammation.

With respect to the treatment of bronchogenic cyst, surgical interventions are recommended to establish a definitive diagnosis, alleviate any symptoms and prevent complications involving especially secondary respiratory infection and the risk of rupture, although the majority of symptoms are not found. Generally, the incidence of malignancy in retroperitoneal bronchogenic cysts is lower than that in thoracic ones⁹⁾, but Sullivan et al.¹⁰⁾ reported a case of retroperitoneal bronchogenic cyst with malignant neoplasm. With the possibility of malignancy taken into account, surgical interventions are an appropriate treatment for retroperitoneal bronchogenic cysts.

Since Tokuda et al.³⁾ initially reported on the surgical treatment with laparoscopic procedures for a retroperitoneal bronchogenic cyst in 1997, there have been only six cases treated with transperitoneal laparoscopic or retroperitoneoscopic surgery³⁻⁶⁾. In the present attempt we successfully performed the operation with five trocars and without any serious operative and postoperative complications associated with our present endoscopic procedures unlike in the previous reports³⁻⁶⁾ showing hemorrhage and/or injury of surrounding organs. From the cyst which we removed surgically, no bacteria were detected, but the liquid contents became turbid. The cyst caused secondary infection and inflammation, and this was suspected as the cause of the low-grade fever. In fact, the fever was promptly alleviated.

Retroperitoneal bronchogenic cyst is a rare congenital anomaly, and even imaging examination rarely reveals a typical cystic pattern. We recommend laparoscopic surgery for the treatment of retroperitoneal small masses, including bronchogenic cysts, because it is a less invasive and safer procedure and a more reliable diagnostic procedure than needle biopsy. Surgical intervention was inevitable in the present case because no preoperative definitive imaging diagnosis was possible.

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和文抄録

腹腔鏡下に摘除した後腹膜気管支原性嚢胞の1例

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症例は39歳の男性。微熱精査中に後腹膜に腫瘤を指摘され、当院を受診となった。腹部CT、腹部超音波断層法において左副腎近傍に35 mm大の均一な充実性腫瘍が認められた。血液生化学検査ではWBC 9,700/ μ L, CRP 1.7 mg/dlと軽度の炎症所見を認めたが、内分泌学的検査では血中、尿中カテコールアミンおよびアルドステロン、コルチゾールともに基準値範囲内であった。炎症所見が軽度なこと、内分泌学的に

も過剰なホルモン産生は確認されなかったため、経過観察を行っていたが1年後の腹部CTでは約10 mmの増大また微熱も持続していた。発熱の原因が後腹膜腫瘤である可能性も否定できないため、腹腔鏡下に後腹膜腫瘍の摘除を行った。病理組織学的に腫瘍は気管支原性嚢胞であった。

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